Auto-amputated ovary- rare cause of intestinal obstruction in a neonate: A case report

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KEYWORDS
Neonate,
Intestinal obstruction,
Dermoid cyst,
Ovary,
Ovarian cyst

ABSTRACT
Background: Congenital ovarian cysts rarely auto amputate. An unusual complication of this event is secondary intestinal obstruction. The mechanism may be consequent to a direct pressure effect or to adhesions induced by the cyst.

Case Presentation: A neonate presented with an abdominal mass and intestinal obstruction. On exploration, she had a mass hanging over the small intestine and one ovary was absent. Mass was excised and it turned out to be an auto-amputated ovary

Conclusion: Although it is a rare pathology in neonates, surgeons must suspect it, particularly if an ovary is absent.

INTRODUCTION
Abdominal masses in neonates are not infrequent, and fortunately most (85%) are benign. These most frequently arise from the Urinary (50%), genital (15%), gastrointestinal (15%), and hepatobiliary systems (5%). [1] Ovarian cysts account for a third of the masses originating from the internal genital tract. Very rarely, ovarian cysts may undergo torsion, auto-amputate, and float freely in the abdominal cavity or attach to another organ. [2] This might result in intestinal occlusion necessitating surgical intervention with a few cases described in the neonatal period. [3] Herein, we report a neonate with an auto-amputated ovary, leading to intestinal obstruction.

CASE REPORT
A 4-day-old, full-term female, weighing 2500 grams, presented with features of bilious vomiting and abdominal distention. Antenatal scans were normal with no fetal abnormality detected. On physical assessment, the abdomen was distended with obstructive bowel sounds and a soft mass was palpable in the hypogastrum. X-ray abdomen showed small bowel obstruction. Ultrasonography demonstrated a 44mm x 48 mm echogenic lesion in the right pelvis without vascular signals. CT scan demonstrated a well-defined, 4.6 x 3.8 x 4.8 cm, rounded, peripherally enhancing thick-walled cystic lesion in the right lower abdomen of unknown origin.

The patient was prepared for surgery and on exploration, a discolored, necrotic cyst was immediately apparent (Fig.1). This was free of a vascular pedicle and firmly attached to a segment of the terminal small bowel which was obstructed (Fig.2). Her uterus and left ovary were normal, but the right ovary was not evident. This mass was presumed to be an auto-amputated cyst arising from the right ovary and once freed from the ileum, was excised. The patency of the
intestine was confirmed suggesting that the obstruction was secondary to a mass effect.

Figure 2: Mass causing intestinal obstruction due to pressure on its lumen.

The postoperative recovery was smooth. Histopathological examination of the lesion confirmed it to be a dermoid cyst of ovarian origin containing ectodermal and mesodermal elements.

DISCUSSION

The auto-amputated ovary is a rare entity with a reported incidence of 1 in 11421. The first case was reported by an Italian anatomist Giovanni Morgagni in 1748 in a postmortem. Subsequently, almost 50 cases have been reported in postmortems. In 1945 the first case was described in a child. More recently, Focseneanu et al. in a comprehensive literature review described a total of 94 cases in the pediatric population. [4]

The etiology of this entity is unknown but thought to be consequent to chronic torsion with necrosis and separation of the vascular pedicle. Another theory advanced is the congenital absence of the adnexa; however, most authors find this implausible. The free-floating cyst is then liable to attach to any organ including the omentum. [4] Alternatively, it might simply involute or calcify. [4, 5] There is another theory stating that combined tubo-ovarian torsion due to an enlarged ovary. [6] And this may have a pressure effect over the ileum, like in our case.

Presenting features in children are; abdominal pain, a palpable mass, or sometimes an incidental discovery. [5, 6] Jeanty et al. reported a case of ovarian cyst that led to intestinal obstruction in a neonate and as part of a more comprehensive review identified 19 cases with intestinal obstruction. [3] However, intestinal obstruction due to an auto-amputated ovary is a rare event. [4] Detection of ovarian torsion is difficult in children and practically impossible in neonates and is usually diagnosed only after exploration. The differential diagnoses in neonates include; mesenteric and duplication cysts. [5]

Ultrasound is the most useful investigatory modality but is not always helpful in detecting ovarian torsion in neonates. Chinchure et al. had found in their retrospective study that cysts with ‘fish-net appearance,’ fluid-debris level, and cysts with an echogenic nodule, favor torsion. [7] CT scan or magnetic resonance imaging may be considered when ultrasound findings are doubtful. [2] Confirying a free-floating cyst with any degree of certainty will be difficult with all the above radiological tools.

The antenatal sonography might be useful in detecting an ovarian cyst although this was not the case with our patient. Focseneanu et al. reported that 93% of the patients with free-floating ovaries had an abdominal cyst visualized on antenatal scans. This is a testament to the rapid advances being made in the field of antenatal scanning where ovarian or other intra-abdominal cysts are rarely missed. [4, 7]

A general rule for a simple ovarian cyst being less than 4cm is observation and regular sonography. However, if the cyst is complicated or getting symptomatic, exploration may be necessary. Considering the widespread use of laparoscopy among neonates, most recent literature shows the advantage of this technique. [8] The most employed treatment for auto-amputated ovarian cyst is surgical excision, mainly if it is symptomatic or causing an intestinal obstruction. [5, 9] In this case, the child had intestinal obstruction with an abdominal mass of unknown origin, so the only option was exploration. Although this cyst was attached by a fibrous cord to the uterus, we were unable to label it an ovary because of its color and location, so we excised it. Non-surgical and expectant treatment has also been reported in newborns with the prenatal diagnosis of auto-amputated ovaries. [10]

Although it is challenging to make the diagnosis of auto-amputated ovary preoperatively, in case of non-visualization of one ovary along with an abdominal mass, it must be kept in the differential diagnoses list. The management may vary with the presentation of the neonate and if there is some ovarian tissue visible, it may be left there for observation and expectant management.

Acknowledgements: Nil

Conflict of Interest: Authors have no conflict of interest.

Source of Support: Nil

Consent to Publication: Author(s) declared taking informed written consent for the publication of clinical photographs/material (if any used), from the legal guardian of the patient with an understanding that every effort will be made to conceal the identity of the patient, however it cannot be guaranteed.

Author Contributions: Author(s) declared to fulfil authorship criteria as devised by ICMJE and approved the final version.
REFERENCES